



Contralateral Perinephric Urinoma Complicated Retrocaval Ureter Repair: A Surgical Trap of Pyelic Fusion Anomaly

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ABSTRACT

Aim: To present a case of pyelic fusion anomaly with orthotopic kidneys which is extremely rare. For maximum and only, it was reported twice in the literature. Its diagnosis may be unpredictable and it may predispose to major surgical complications.

Case Report: An 8-year-old child was diagnosed to have a retrocaval ureter which was repaired through a flank incision. A striking finding of a cut and retracted tubular structure was noted during ureteral dissection. Postoperative contralateral perinephricurinoma developed and drained percutaneously. Then, two surgical explorations were done on the left kidney without detection of a cause for urinary extravasation. The left ureter was atretic during explorations. Kidney autotransplantation and ileal reconstructions seemed difficult solutions and nephrectomy was the final step.

Discussion: Pyelic fusion anomaly was reported, mainly, in association to renal fusion anomalies like horseshoe and crossed ectopic kidneys. However, it is the first time to report a case of retrocaval ureter in association to pyelic fusion and normally-located and positioned kidneys. Contralateral urinoma after repair of a retrocaval ureter was a striking complication referred to an underlying crossing left-to-right anomalous pathway. This case represented a surgical trap, because it is an extremely rare and unpredictable association of renal anomalies.

Conclusion: Pyelic fusion with orthotopic well-positioned kidneys is an extremely rare renal anomaly. Its association to retrocaval ureter makes it extremely unpredictable, especially, when it is undiagnosed preoperatively.

Key Words: Pyelic fusion, Retrocaval ureter, Surgical trap, Urinoma

INTRODUCTION

Although fusion anomalies of the kidney are rare, they are well-known urological lesions, especially the horseshoe kidney. It may associate to the extremely rare anomalies of pyelic and ureteral fusion[1, 2]. The latter may occur, also, with crossed ectopic kidneys[3] or other extremely rare fusion anomalies[4]. However, pyelic fusion anomaly in non-fused orthotopic kidneys is exceedingly rare and hardly detectable in the literature[5, 6]. Previously unreported anatomical anomalies may predispose to major surgical complications like what happened in the following case. So, it is practically attractive and configures a lesson that may deserve to be exposed for learning purposes.

CASE REPORT

An 8-year-old boy presented by mild abdominal pain of many months duration. Abdominal ultrasound revealed moderately obstructed right kidney and normal left side. Plain Kidney-Ureter-Bladder film showed no stones or other abnormalities. Urine analysis parameters were normal and serum creatinine was 0.6 mg/dl. Intravenous urography revealed moderate right hydronephrosis and the upper ureter was dilated with S-shape course consistent with a retrocaval ureter. Both kidneys were well-rotated and the left one was not obstructed, slightly larger in size, but without definite duplication of the pelvicalyceal system. Preoperative routine work up was unremarkable.

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The child was admitted to our department for surgical repair and explored through the right extraperitoneal flank incision. During ureteral dissection at the retrocaval part, the assistant surgeon declared that a small tubular structure was seen cut and retracted within the retroperitoneal tissues. Accordingly, the main surgeon examined the part of upper ureter and renal pelvis and found nothing abnormal. So, he proceeded and completed the steps of the ureteral repair on a JJ stent. The wound was closed on a tubal drain. After 3 days the patient had fever and left side abdominal pain. Abdominal ultrasound revealed a large left perinephricurinoma which was drained percutaneously. Right side drain was draining nothing, so it was removed on the fifth day.

On the 10th day, a new intravenous urography study was requested and revealed fine right kidney with only residual dilatation on JJ stent. The left kidney was normal in shape and not obstructed, but with perinephric extravasation of the dye. Surgical exploration of the left side was done, but surgeons could not define the source of urinary leakage. The left ureter was incised and found atretic at multiple levels with intra-renal small renal pelvis. Formal nephrostomy tube was inserted and the wound was closed for further evaluation. However, urinary leakage continued. So, second exploration by more senior urologists was done after further 3 weeks. However, they found nothing more and, as a reflex, decided nephrectomy. Strikingly, leakage stopped by removal of the left kidney and the patient discharged and the JJ stent was removed eight weeks from the primary surgery.

DISCUSSION

An extremely rare urological anomaly may represent a surgical trap for the urologist when it is unnoticed preoperatively[7]. In the current case, the surgical trap happened and resulted in a contralateral perinephricurinoma as an unpredictable complication for repair of retrocaval ureter. Visualized cut tubular structure during dissection of the right upper ureter and left ureteral atresia on exploration of the left side were definite findings reported by the surgeons. Proposed right side causes for urinoma like an injured duplicated ureter or upper calyceal infundibulum are not accepted, because the urinoma is, simply, on the left side and the right side was free, postoperatively. Also, suggestion of spontaneous rupture of the left kidney is not valid, because the latter was not obstructed at any stage before or after interventions. The only explanation is a congenital left-to-right connection between the pelviureteral segments. Although it is extremely rare, pyelic fusion anomaly with normally-ascended kidneys should be considered. This case represented a surgical hallmark and surprise in our records for many years. However, the suggestion of an underlying pyelic fusion is plausible after reporting of two similar cases in the literature by Enganti et al. [5] and Bhat et al. [6] which encouraged me to report

this unique case which occurred many years ago. The similarity to these cases was in regard to the position and location of the kidneys. The differences, however, were presence of solitary ureter and multiple anomalies in Enganti et al. case and the incidental discovery in Bhat et al. case. Moreover, in the current case and to the first time, retrocaval ureter was a major component. In contrast to the current case, retrocaval ureter usually manifests in adulthood age of 30 – 40 years [8].

Novel imaging techniques might facilitate the diagnoses of the reported cases. However, at the time of presentation of this child, the available employed tools were abdominal ultrasonography and intravenous urography as the best for retrocaval ureter anomaly [8]. They could not diagnose the associated anomaly, preoperatively.

Urinoma is an encapsulated urinary collection which is contained within a thick membrane formed by extensive fibrous reaction of the tissues to the extravasated urine[9]. In the current case, it could be one of the factors hindered the detection of the transected fusing segment during the repeated surgery. Uniquely, the urinoma was contralateral to the primarily diagnosed pathology and the primary surgical intervention.

The decision of nephrectomy could be questionable, but it was a situational reflex. Moreover, the surgeons were reluctant to more complicate this difficult situation by alternatives (autotransplantation or ileal ureteral replacement) which seemed relatively difficult.

To the best of my knowledge, the current case is the first in the literature to report a contralateral urinoma formation associated to retrocaval ureter repair due to an underlying pyelic fusion with well-rotated orthotopic kidneys. At the time where the surgeries for this case were done, it was definitely previously unreported and unpredictable anomaly. Now, however, it is the third reported case of pyelic fusion anomaly with orthotopic well-rotated kidneys.

CONCLUSION

Pyelic fusion with orthotopic well-rotated kidneys is an extremely rare reported urological anomaly. Moreover, its association to retrocaval ureter makes the condition very hardly detectable. It is a surgical trap that deserves declaration to the urologists to help prevent undue major complications in the similar situations.

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Ethical approval:

This case study was approved by the ethical committee of the Faculty of Medicine in Assiut University. Informed consent was taken from the patient's father.

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